Neonatal Bilateral Testicular Torsion: A Plea for Emergency Exploration

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Purpose: Bilateral testicular torsion is a rare condition. Most authors present single case reports. Therefore, the clinical and surgical aspects of bilateral torsion in a neonate have not been subjected to detailed analysis. We performed a retrospective analysis of our experience in the management of bilateral perinatal torsion as well as a collective review of the medical literature.

Materials and Methods: All cases of neonatal testicular torsion managed at our neonatal surgical center during the last 2 decades (1986 to 2005) were reviewed, and 3 cases of bilateral torsion were identified. In addition, 45 neonatal cases of bilateral torsion were found through the literature search. In all cases data regarding clinical presentation, imaging studies, surgical management, intraoperative and pathological findings, and final outcome were analyzed.

Results: Synchronous torsion occurred in 32 of 48 newborns (67%), while asynchronous pathology was reported in 16 (33%), including the 3 presented in this report. All except 1 patient were full-term newborns with normal or above average birth weight. Difficult delivery was noted in 33% of the cases. Despite prompt surgical intervention in 46 infants, the salvage rate was low, with arterial flow confirmed postoperatively in only 3 gonads (3.1%). Four gonads in 3 additional patients were reported to be of normal size on followup.

Conclusions: Asynchronous torsion is not as rare an event as previously reported, and it may pose a diagnostic challenge. In the majority of these cases torsion of the left testis seems to occur later than torsion of the right testis. The role of imaging studies in newborns with bilateral torsion seems to be limited, especially in cases of asynchronous pathology. Urgent bilateral exploration is strongly advised in all newborns presenting with either unilateral or bilateral torsion. Such policy carries diagnostic, potential therapeutic and prognostic implications.

Key Words: spermatic cord torsion; testis; infant, newborn

Testicular torsion is an acute surgical emergency. It may occur at any age but peaks in the neonatal and adolescent age groups. A policy of emergent surgical intervention has been uniformly accepted in older boys with testicular torsion.[1,2] Management of neonatal torsion has not been associated with such uniformity. The necessity of emergent exploration and its timing remain the most controversial issues. The poor salvage rate associated with a natal diagnosis seems to influence many surgeons to postpone the operation. Others claim that surgical intervention needs to be performed on an emergency basis as the only reliable method for diagnosis, potential reversal of the ischemic event and fixation of the contralateral testis. The latter strategy has been recommended for newborns presenting with unilateral torsion.[3–5]

There is a small subset of newborns presenting with bilateral torsion. Although a rare event, it carries an immediate risk of anorchia.[6,7] No more than 50 newborns have been reported to date with bilateral testicular pathology. Due to limited clinical experience, bilateral torsion has not been a subject of detailed analysis. We report 3 additional cases and discuss the clinical and surgical aspects of this condition.

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MATERIALS AND METHODS

We reviewed the records of all newborns with testicular torsion treated within the first 30 days of life at our institution between 1986 and 2005. The medical notes were analyzed and cases of bilateral torsion were identified, and form the basis of this report. The relevant data regarding clinical presentation, surgical management, and intraoperative and pathological findings were studied in each case. A detailed search of the medical literature dealing with the subject of neonatal bilateral testicular torsion was conducted to gain more objective insight into this pathological condition.

RESULTS

During the last 2 decades 58 newborns with testicular torsion were treated at our institution. In all patients a final diagnosis of testicular pathology was confirmed intraoperatively. The policy of prompt operative intervention with inspection and fixation of the contralateral testis, as soon as suspicion or clinical diagnosis of torsion had been established, was consistent throughout the study period. All 58 boys had extravaginal torsion.

Bilateral testicular torsion was diagnosed in 3 patients (5.2%). All were full term and were born after an uneventful pregnancy. In 2 of these patients scrotal pathology was noted on the initial postnatal examination. One patient presented with an enlarged, hard and nontender left testis, 1

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had painless enlargement of both gonads and 1 had a firm, nontender right scrotal swelling noted on day 2 of life. Both patients presenting initially with unilateral scrotal findings underwent scrotal ultrasound. In 1 patient it revealed a surprisingly similar appearance of both testes presenting as a mass of mixed echogenicity with loculated fluid. In the other patient ultrasound performed at the referring neonatal unit was inconclusive.

The 3 patients with bilateral torsion underwent emergent scrotal exploration at 18, 26 and 72 hours of life. Complete infarction of the gonads was noted intraoperatively in 2 patients with asynchronous torsion, and both underwent orchiectomy. Pathological examination confirmed ischemic necrosis of the affected testes. The findings of fibrosis and calcification of infarcted tubular structures were suggestive of long-standing vascular accident. The remaining newborn had synchronous torsion with marked ischemia and congestion of both gonads. Incisional bilateral biopsy was performed and the testes were fixed. Pathological examination showed extensive hemorrhagic infarction with little viable tissue remaining. At 6-month followup both gonads were atrophic.

DISCUSSION

Bilateral testicular torsion is a rare neonatal condition. Its true incidence is difficult to assess objectively, since most authors have presented isolated cases. We noted a frequency of 5.2% during a period of 20 years. The rarity of this condition is reflected in the fact that we were able to collect data for only 45 newborns from the medical literature.

Perinatal torsion of the testis has been arbitrarily defined as occurring prenatally or postnatally within the first 30 days.[4] The exact timing of this disastrous event cannot be objectively determined, but it has been assumed that the majority of perinatal torsions take place in utero.[4,6] Pathological examination of the testicular tissue may give a further clue for confirming the course of events in our 2 patients with evidence of calcification and fibrosis. Such findings have been reported by many authors.[6–8] From this point of view, unilateral and bilateral torsion seem to share similar pathological features.

The evident proof for prenatal occurrence of testicular vascular catastrophe is based on prenatal diagnosis. Among 48 newborns prenatal ultrasound had demonstrated hydrocele and heterogeneous structure of the affected testes in 4. Three of these patients presented postnatally with bilateral scrotal swelling, and in the remaining patient torsion of the contralateral nonsymptomatic testis was noted intraoperatively.[5,6,9,10]

The etiology of perinatal bilateral torsion remains an unresolved issue. Review of 48 cases, including our series, revealed that all but 1 involved extravaginal torsion.[11] With the exception of a 32-week newborn, all patients were born at term.[12] A total of 20 patients had a birth weight exceeding 3,500 gm, while 8 weighed less than 3,500 gm. In 20 cases data regarding birth weight were not included.

An above average birth weight is the most striking feature in neonatal torsion but its true significance remains unclear. Some authors have noted a high incidence of difficult delivery among these newborns.[11,13] It may be speculated regarding whether a difficult delivery has any potential impact on testicular pathology or is simply related to greater than normal weight in a male fetus. If the former hypothesis were true, it would serve as an explanation for intrapartum and postnatal events only. Among 33 newborns in whom obstetrical data were presented 11 (33%) were born by either cesarean section or nonspontaneous vaginal delivery.[5,11,13,14] It has been hypothesized that perinatal torsion is a result of lack of or loose scrotal attachments of the testis, with a subsequent predisposition to rotate along the long axis of the spermatic cord.[12,15] Were this anatomical abnormality of the testis the sole etiological factor, it would be difficult to explain why testicular torsion occurs almost exclusively in full-term babies.

Bilateral testicular torsion may be synchronous or asynchronous. Surprisingly, despite widely presented opinions of extreme rarity of the latter form, we were able to document an incidence of 33% of all bilateral torsions.

In 17 of 32 patients with synchronous torsion typical clinical symptoms of scrotal swelling, induration and skin discoloration were noted at birth.[1,6,7,9–11,13–16] In 5 additional patients, including 1 of ours, both testes were firm and nontender on routine examination.[8,13,17,18] Among 6 newborns in whom symptoms were recognized within first 12 hours of life 3 had normal scrotal anatomy immediately postnatally.[11,13,16,19] In 4 cases clinical diagnosis of testicular pathology was made at 2, 4 and 10 days of life.[1,11,20] In 1 newborn a clinical diagnosis of epididymo-orchitis was made, and atrophy of both testes was noted at age 16 years during exploration for undescended testes.[18]

Imaging studies were performed in 11 newborns. In 5 of these patients ultrasound was the only investigation used, while in 6 ultrasound was supplemented with Doppler imaging and/or radionuclide study. In 1 boy an initial Doppler examination showed increased pulsations, while subsequent scintigraphy demonstrated avascularity of the testicular tissue.[17] In single cases results of ultrasound and radionuclide scan were described as inconclusive.[7,10] All but 2 patients in this group were subjected to surgical exploration.

Among 30 patients 20 were operated on within the first 24 hours of life. Seven patients were older than 3 days when subjected to scrotal exploration. Surgical management consisted of bilateral orchiopexy in 12 newborns, bilateral orchiectomy in 13 and right orchiectomy with left orchiopexy in 4. Among 16 patients in whom orchiopexy was carried out followup observation was available in 8. Atrophy of the testes was noted in 3 patients, while smaller and firm gonads were reported in 3.[11,16] One patient had 2 testes of normal size at age 16 weeks.[11] In 1 case histological examination of testicular biopsy revealed surviving Leydig cells despite the intraoperative appearance of necrosis.[6] Therefore, it may be concluded that none of the patients with synchronous bilateral torsion showed objective proof of testicular salvage, regardless of the timing of surgical intervention.

Clinical presentation of asynchronous testicular torsion is much more variable.[2–5,11–14,20] Initial symptoms of unilateral scrotal pathology may be associated with normal anatomy of the contralateral scrotum or late presentation of contralateral disease following an asymptomatic period of variable duration after birth. The first symptoms were noted postnatally in 13 of 16 patients, and in 1 each at 22 and 28 hours, and day 2 of life. There was an equal distribution with regard to initially symptomatic scrotal side. At the time of diagnosis the contralateral testis was reported as normal in